Case Report

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Cystocutaneous fistula of the left lobe of liver: an extremely rare presentation of hydatid liver cyst

Suryakanth D. Mandolkar^{1*}, Ramakanth B², Anil Kumar PL³, Sudarshan GT³

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*Correspondence:

Dr. Suryakanth D. Mandolkar,

E-mail: drsuryakantsurgery@gmail.com

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ABSTRACT

We report a case of spontaneous cutaneous fistula formation consequent to an infected hydatid cyst of the left lobe of the liver in a 25 year old female patient. This is a very rare natural presentation of the disease and owing to improved diagnosis and therapy, complications of the disease is also rare. Ultrasound of the abdomen revealed a cystic lesion on the left lobe containing multiple daughter cysts. The patient underwent partial pericystectomy and complete excision of the fistula. The importance of correct diagnosis and the requirement for fast and radical surgery for the complete excision of fistula and cyst evacuation in hydatid disease of the liver is stressed in this report.

Keywords: Fistula, Cystocutaneous, Rupture, Hydatid liver disease, Surgery, Rupture

INTRODUCTION

Cystic Echinococcosis (CE) which involves the formation of echinococcal cyst in any tissue or organ of the body is caused by Echinococcus granulosus infection. The most common organ affected by the disease is the liver where over 50 to 70 % of the infections occur and another 20 to 30 % of the infections affect the lungs. The disease is generally asymptomatic until it is diagnosed incidentally or when infection or rupture of the cyst occurs. The cysts can rupture into any body cavity including peritoneal cavity, gastrointestinal tract, pleural cavity and bile ducts. In case of liver hydatid disease, the left hepatic lobe is affected only in 20% of the cases and spontaneous cysto-cutaneous fistula is a rare presentation with only a very few cases reported in the literature. 3-5

CASE REPORT

A 25-year-old female patient was presented to the emergency department with a swelling in the left lower chest and left hypochondriac region. The patient was unmarried and came from a rural region and reported a continuous and throbbing pain in the area for the past one-month. On examination, there was a palpable and diffuse lump over the left hypochondrium and left lower chest with cystic consistency. Further, the pain was also associated with a history of fever, chills and rigors, which subsided following spontaneous rupture of the swelling through the left lower chest forming a fistula, discharging the cystic elements. The patient was also anemic and blood test revealed polymorphic leucocytosis. Ultrasound of the abdomen revealed an echo texture cystic lesion 11 x 10 x 10 cm in size extending from the left lobe of the

¹Department of General Surgery, Raichur Institute of Medical Sciences, Raichur-584102, Karnataka, India

²Department of General Surgery, BLDEA's Shri BM Patil Medical College Hospital & Research Centre, Bijapur-586103, Karnataka, India

³Department of Surgery, BLDEA's Shri BM Patil Medical College Hospital & Research Centre, Bijapur-586103, Karnataka, India

liver. It was interspersed with multiple daughter cysts discharging cystic elements through the fistula from the medial to the left mid clavicular line located in the 8th intercostal space (Figure 1). Hepatomegaly was evident and no other masses were palpable. The lesion also appeared to be communicating with left hypochondriac skin surface through the intercostal space. X-ray chest with upper abdomen disclosed an erosion of the 9th rib and tenting of the left hemi diaphragm with clear left costophrenic angle. Based on the clinical presentation and ultrasound findings, the diagnosis of hydatid cyst of the liver with spontaneous cutaneous fistula was established.



Figure 1: Daughter cyst extruding from the fistulous opening.

Due to the presence of dense adhesions between the cyst and the anterior wall of the abdomen, laparotomy was performed with a left sub costal incision. A thorough intraoperative examination of the abdominal cavity showed a large cyst localized to the left lobe of the liver with a fistulous communicating tract to the external surface (Figure 2). Open cyst evacuation with partial peri-cystectomy was performed. The residual cystic cavity was cleared of its contents and irrigated with diluted povidone iodine solution. The fistula tract was excised and the cavity was drained externally.



Figure 2: Intra operative photograph showing the cyst cavity communicating to the external surface.

The postoperative course was uneventful and the patient underwent one course of antiparasitic therapy with albendazole 10 mg/kg for duration of two months. At 4 months follow up, the patient was doing well and was free of symptoms.

DISCUSSION

Hydatid disease is endemic in developing countries and cattle rearing regions of the world. The most common causative organism is Echinococcus granulosus where the infected humans are median hosts in the life cycle of the parasite.⁶

Infection of the left lobe of the liver is relatively rare compared to right lobe infections. Albiet, the disease is of benign nature, and asymptomatic in most of the cases, complications occur rarely due to spontaneous rupture into the biliary tract which has a reported incidence of 5-10%. It can also rupture into the peritoneal cavity, bronchus and rarely into hollow viscus.⁷

The occurrence of spontaneous cysto-cutaneous fistulae is a highly rare complication of the disease condition. The cysts located in the liver margins could grow progressively and exhibit a tendency to erode the adjacent tissues and organs. With time, the damage is accentuated through muscle penetration, subcutaneous rupture and by formation of fistula. As seen in the case of our patient, the cysts from left lobe affect the anterior wall of the abdomen.

Cystic echinococcosis derives special clinical interest owing to the fact that it is difficult to diagnose and the complications could bring fatal and life threatening consequences for the patient. Surgery is the appropriate method of choice for such complicated cysts and complete resection of the cyst including the fistula tract is indicated as done in the case of the patient presented in this report. Radical surgery or total pericystectomy, conservative surgery or open cystectomy and simple tube drainage are the major surgical options for infected and communicating hydatid cyst. Other procedures include 'PAIR' and laparoscopic procedure.

The incidence of the disease could be decreased by creating awareness on the preventive measures, public education and improved health care and veterinary measures. As early detection of the disease is facilitated by means of advanced imaging techniques, complications of the disease occur rarely. Only less than five cases of hydatid liver disease with spontaneous cutaneous fistula have been reported in the literature. Due to its infrequent occurrence misdiagnosis of the condition is possible and the present report emphasizes the likelihood of such a diagnosis. Moreover, the need for a speedy intervention involving complete excision of the fistula is highlighted in order to prevent secondary occurrences and avoid post-operative complications.

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